

proliferative and cytotoxic capacity *in vitro* against cultured lymphoma cells⁷. This immature or abnormally developed immune system may predispose to the development of malignant change, as has been found in diseases of postulated immunological aetiology such as thyroiditis. Another suggestion is that the immunological disturbances in coeliac disease may activate oncogenic viruses⁶.

T cell lymphoma most often presents as tumour masses in the small bowel, and neurological presentation with lymphomatous meningitis has not previously been reported. The possibility of lymphoma should be considered in coeliac patients who deteriorate neurologically for no apparent reason.

REFERENCES

- 1 Gull W. Fatty stools from disease of the mesenteric glands. *Guy's Hosp Rep* 1885;1:899
- 2 Cooper BT, Holmes GKT, Ferguson R, Cooke WT. Celiac disease and malignancy. *Medicine (Baltimore)* 1980;59:249–61
- 3 Gough KR, Read AE, Naish JM. Intestinal reticulosis as a complication of idiopathic steatorrhoea. *Gut* 1962;3:232
- 4 Harris OD, Cooke WT, Thompson H, Waterhouse JAH. Malignancy in adult celiac disease and idiopathic steatorrhoea. *Am J Med* 1967;42:899
- 5 Posner JB, Chernik NL. Intracranial metastases from systemic cancer. In: Schoenberg BS, ed. *Advances in Neurology*. New York: Raven, 1987:575–86
- 6 Swinson CM, Coles EC, Slavin G, Booth CC. Coeliac disease and malignancy. *Lancet* 1983;111–15
- 7 Spencer J, MacDonald TI, Diss TC, Walker SJA, Ciclitira PJ, Isaacson PG. Changes in intraepithelial lymphocyte subpopulations in coeliac disease and enteropathy associated T cell lymphoma (malignant histiocytosis of the intestine). *Gut* 1989;30:339–46
- 8 Bayless TM, Kapelowitz RF, Shelley WM, Ballinger V, Hendrix TR. Intestinal ulceration, a complication of coeliac disease. *N Engl Med* 1967;276:996

Solitary cellular schwannoma presenting with haemothorax

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Haemorrhage from intrathoracic neurogenic tumours is extremely rare, and most cases are reported in patients with multiple tumours associated with Von Recklinghausen's disease^{1–4}. We record here a case of haemothorax caused by rupture of a solitary cellular schwannoma.

CASE HISTORY

A man aged 27 was admitted to his local hospital with left-sided loin pain and a week's history of upper respiratory tract infection. The pain had developed gradually and was made worse by movement, including inspiration and coughing. He had no medical history of note, but mentioned that he had received a blow to the left side in the course of a 'play fight' four days previously. On examination he was afebrile and stable. The chest wall was not tender. Decreased air entry was noted at the base of the left lung and there was tenderness in the left hypochondrium without

guarding or rebound pain. The white cell count was $13.8 \times 10^9/L$. Urine microscopy was normal. Chest radiograph showed a rounded opacity projected over the left hilum together with an area of consolidation in the left lower lobe. In addition, there was erosion of the undersurface of the left seventh rib (Figure 1). An abdominal radiograph appeared normal.

He was admitted for further imaging investigations and antibiotics were prescribed for a presumed lobar pneumonia. Although initially stable, he became pale and lethargic, with an unproductive cough. Computerized tomography (CT) demonstrated the presence of a large left pleural effusion with complete collapse of the left lower lobe. An intercostal chest tube was inserted and three litres of bloodstained fluid were drained. The fluid contained no organisms and had a protein count of 49 g/L. Post-drainage CT showed a well-defined loculus in the left paravertebral gutter, with the appearance of fluid attenuation but containing some internal structures (Figure 2). Extensive atelectasis was noted in the left lower lobe. Intrathoracic neoplasia was considered at this stage but ultrasound-guided biopsy of the loculus showed fibroadipose tissue, granular tissue and blood clot. There was no evidence of neoplasia. Bacteriological culture showed no growth. A thoracic surgical opinion was sought and it was felt that the patient needed operative intervention, both to achieve satisfactory re-expansion of the lung and, in the light of the radiological findings, to investigate the source of bleeding.

Left video assisted thoracoscopy via three ports revealed a loculated haemothorax with a dense fibrous cortex restricting the lower lobe. An axillary muscle-sparing incision was performed via the upper two port site incisions. A litre of blood was aspirated from the pleural cavity, allowing identification of a $6 \times 5 \times 4$ cm encapsulated

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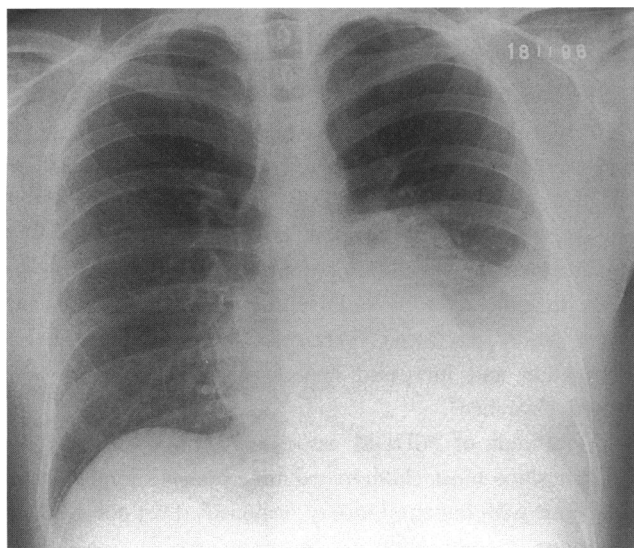


Figure 1 Chest radiograph at initial presentation

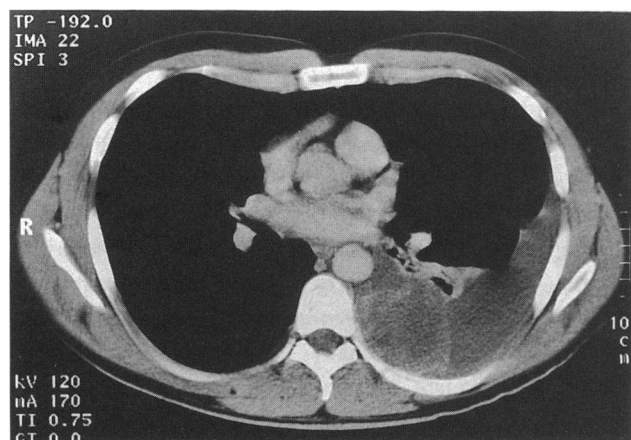


Figure 2 Post-drainage computerized tomography showing loculated mass in left paravertebral gutter

mass arising from the 7th intercostal nerve on the posterior aspect of the chest wall. The mass was easily excised, with proximal and distal division of the intercostal nerve, and a full decortication of the lung was carried out. At microscopy, the tumour was cellular with fascicular architecture. The cells were wavy, spindle-shaped and mildly pleomorphic. Moderate mitotic activity and patchy necrosis were noted, along with extensive intratumoural haemorrhage (Figure 3). Haemosiderin deposition was noted. Staining for S-100, a neural marker, was strongly and diffusely positive, with epithelial and smooth muscle markers negative. In view of its encapsulation and relation to nerve, together with its diffuse S-100 staining, a diagnosis of cellular schwannoma was made. Postoperative recovery was uneventful and the patient was discharged after five days.

COMMENT

Haemothorax from solitary cellular schwannomas has not to our knowledge been reported previously, the closest



Figure 3 Cellular spindle cell neoplasm showing extensive intratumoural haemorrhage (haematoxylin and eosin $\times 25$)

comparison being the case of a ruptured feeding artery to an intrathoracic neuroma⁵. In a series of 57 cellular schwannomas, only 3 were in the chest wall and none presented with haemothorax⁶. In this case, presentation followed an episode of trauma which most likely caused haemorrhage and subsequent breach of the capsule, but the presence of intratumoural haemosiderin indicates previous spontaneous haemorrhage within the mass which probably weakened the structure of the tumour, predisposing it to rupture.

From a clinical viewpoint, the history of trauma could have contributed to diagnostic delay, but recognition of the implications of rib erosion at first presentation allowed the correct diagnosis to be considered early. Thoracotomy was necessary not only as a diagnostic procedure, but also to enable decortication, ensure completeness of excision and free trapped lung. In haemothorax an underlying neoplasm should always be considered, even when there is a history of trauma.

REFERENCES

- 1 Larrieu AJ, Hashimoto SA, Allen P. Spontaneous massive haemothorax in Von Recklinghausen's disease. *Thorax* 1982;37:151-2
- 2 Wander JV, Das Gupta TK. Neurofibromatosis. *Curr Probl Surg* 1977;14:1-81
- 3 Butchart EG, Grotte GH, Barnsky WC. Spontaneous rupture of an intercostal artery in a patient with neurofibromatosis and scoliosis. *J Thorac Cardiovasc Surg* 1975;69:919-21
- 4 Fuyuno G, Kobayashi R, Iga R, et al. A case of Von Recklinghausen's disease associated with a haemothorax due to a rapidly growing malignant schwannoma. *Jap J Thorac Dis* 1995;33:682-5
- 5 Tanita T, Ohkuda K, Nitta S, et al. A case of intrathoracic neurinoma presenting as hemothorax. *Jap J Thorac Dis* 1981;19:127-30
- 6 White W, Shiu MH, Rosunblum MK, Erlandson RA, Woodruff JM. Cellular schwannoma. A clinicopathologic study of 57 patients and 58 tumors. *Cancer* 1990;66:1266-75